Available online at www.sciencedirect.com

ScienceDirect

Tanta Dental Journal 11 (2014) 109–113

www.elsevier.com/locate/tdj

Heamangioma in parotid gland encroaching on the temporomandibular joint: A case report

S.A. Elsayed*, H.H. Alahmady

Oral and Maxillofacial Surgery, Al Azhar University, Girls Branch, Department of Oral and Maxillofacial Surgery, Al Azhar University for Girls, Cairo, Egypt

Received 3 March 2014; revised 14 June 2014; accepted 15 June 2014
Available online 26 September 2014

Abstract

The majority of swellings in the parotid region are sialadenitis, neoplastic lesion or lymph node. Vascular malformation is a rare cause of swelling of the parotid gland. Parotid heamangioma usually presents as a cystic swelling in the preauricular area but may mimic sialadenitis making the diagnosis difficult. We are reporting, to our knowledge, the first case of parotid gland heamangioma with its clinical presentation, radiological and surgical features which extended medially to the temporomandibular joint. The rarity of such condition in head and neck region and misdiagnosis or failing to diagnose make this case more interesting to the maxillofacial surgeon.

Keywords: Parotid gland; TMJ heamangioma

1. Introduction

The parotid gland is the largest of all major salivary glands. In the adult, the gland is entirely serous in secretion, situated in the space between the posterior border of the mandibular ramus and the mastoid process of the temporal bone. The external acoustic meatus and the glenoid fossa lie above together with the zygomatic process of the temporal bone. On its deep (medial) aspect lies the styloid process of the temporal bone. Inferiorly, the parotid frequently overlaps the angle of the mandible and its deep surface overlies the transverse process of the atlas vertebra. The shape of the parotid gland is variable. Often it is pyramidal in shape, with up to 5 processes. However, on occasion it is more or less of even width and occasionally it is triangular with the apex superiorly. In 20% of subjects a smaller accessory lobe arises from the upper border of the parotid duct approximately 6 mm in front of the main gland. This accessory lobe overlies the zygomatic arch [1].

The gland is surrounded by a fibrous capsule previously thought to be formed from the investing layer

* Corresponding author. Tel.: +20 (002)01121281982.
E-mail addresses: shadiaelsayed25@gmail.com, deepfacial@yahoo.com (S.A. Elsayed).

Peer review under the responsibility of the Faculty of Dentistry, Tanta University

Production and hosting by Elsevier

http://dx.doi.org/10.1016/j.tdj.2014.06.005

of deep cervical fascia. This fascia passes up from the neck and was thought to split to enclose the gland. The deep layer is attached to the mandible and the temporal bone at the tympanic plate and styloid and mastoid processes [2]. Recent investigations suggest that the superficial layer of the parotid capsule is not formed in this way but is part of the superficial musculoaponeurotic system (SMAS) [3].

Hemangiomas are typically found in the pediatric age group. The majority are of the cavernous type and less likely the capillary type. They are typically slow flow lesions and may not be angiographically evident. Of all salivary gland tumors, the vast majority (80%) are found in the parotid gland. The submandibular gland contains approximately 10%, with the remainder in the sublingual and minor salivary glands. Hemangiomas are benign proliferations of vessels closely resembling normal vessels. Their similarity to normal vessels is so great that it is unclear whether they represent vessel malformations, true neoplasms, or hamartomatous overgrowths. Malformations that present at birth or those that appear shortly after birth are all congenital and represent vascular malformations. However, many vascular lesions may be congenital but subclinical at birth, only to appear years later (eg hereditary hemorrhagic telangiectasia and several facial hemangiomas). Indeed, many arteriovenous hemangiomas that emerge in the late teens and early 20s are associated with other abnormal vessels identified only by angiography [4].

2. Case report

A 19-year-old male presented to our outpatient clinic, Department of Oral and Maxillofacial Surgery, Al Azhar University, Girls' branch with complaint of unilateral facial swelling in right parotid region since childhood. The swelling had gradually increased to the present size. The swelling did not show any change in size during meals, bending or straining. There was no history of sudden increase in the size of swelling. The patient had no numbness or any abnormal sensation. Clinical examination revealed a solitary, firm, non-tender swelling of approximately 2 × 2 cm in size with ill-defined margins. It was immobile from side to side and the overlying skin was normal in color, texture and temperature similar to surrounding skin (Fig. 1). FNAB (Fine Needle Aspiration Biopsy) was done which yielded only blood. A computerized tomography (CT) scan was done which demonstrated a large lobulated mass in the superficial lobe of the right parotid gland region measuring about 3 cm × 4 cm

Fig. 1. Preoperative photograph showing swelling in the right parotid region.

(Fig. 2). Even though a history, clinical examination and CT scan, potentiated the impression of parotid benign condition, an MRI was ordered and excision of the mass was planned. At surgery, a well defined, encapsulated, bluish dark colored lesion was seen involving upper part of the parotid gland. With careful dissection, the mass revealed to be attached to the capsule and the TMJ disc and encroaching on the condylar head making obvious discolored condylar head. Sharp dissection was performed to separate the lesion from the lateral side of the articular disc without any risk of bleeding as the mass was intact and surrounded with a thick wall. The facial nerve was not detected in the field and did not need dissection, and the specimen was removed in toto. Histopathological examination of the specimen revealed lobules of variably sized large congested blood filled cavernous spaces with interstitial hemorrhages. The blood spaces lined by flattened endothelium and were surrounded by fibrofatty tissue and foci of preauricular lymphoid tissue aggregates, with no signs of malignancy (Fig. 3). The final histopathological diagnosis was cavernous heamangioma. Post-operative healing period was uneventful. The drain was removed on the 3rd post-operative day and the patient was discharged on the 7th post-operative day after suture removal.

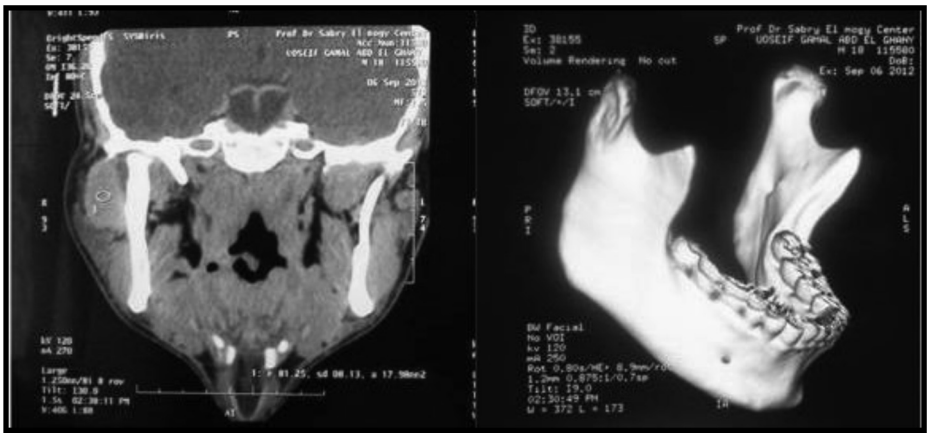

Fig. 2. Computed tomography imaging (soft tissue window) and 3DCT showing a well marginated, homogenous mass in the upper part of the parotid gland.

3. Discussion

The evaluation of a mid-cheek mass represents a significant diagnostic and therapeutic challenge. Benign and malignant lesions in this area may arise from soft tissues of the face, including the skin, lymphatics, skin adnexa, neural, and salivary structures. Benign tumor of mesenchymal origin that involves major salivary gland often poses a difficult diagnostic dilemma [5]. If solely based on clinical features and history, the preoperative assessment may suggest a benign epithelial tumor of salivary gland or salivary calculus disease [6]. Among heamangioma of salivary glands 90% are seen in the parotid gland while the remaining 10% involves the submandibular gland. In the reference series reported by Yadav JS, 2011 [7] from Armed Forces Institute of Pathology; only 1.4% of all salivary gland tumors were found to be benign mesenchymal tumors; out of these, 30% were haemangioma. A probable explanation of this big incidence difference between parotid and submandibular is the lack of a well-defined capsule and the presence of neurovascular structure in the parotid gland [7]. Haemangioma of major salivary gland especially parotid is quite common in pediatric population. Ninety percent of heamangiomas arise in the first three decades of life and are the most common lesions of the major salivary glands during infancy and early childhood. In the present case, cavernous heamangiomas, occur in an adult patient. This goes in accordance with Nagao et al. [8] in a series of 20 cases of cavernous heamangioma of parotid gland; the average age was 26 years (range, 4 months to 50 years). Heamangiomas occur twice as often in females than males and may fluctuate in size with pregnancy and menarche suggesting that the endothelial cells may be quite responsive to circulating hormones [7]. In contrary to the present case, an observed left sided preference had been reported [8]. The diagnosis of haemangioma in salivary glands, especially in adults are quite difficult, as these lesions

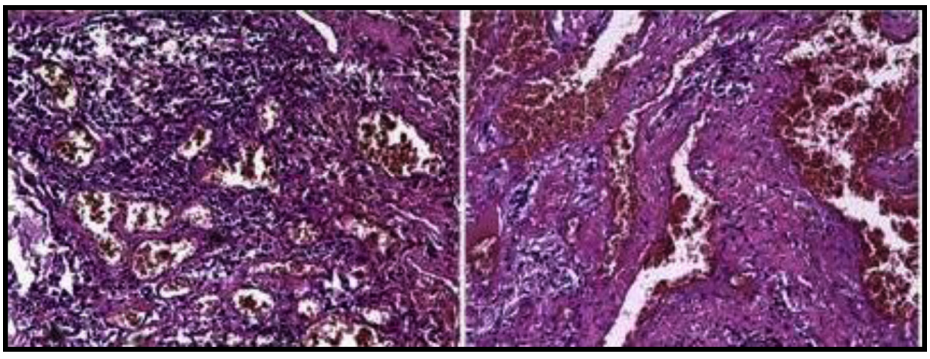

Fig. 3. Histopathology of Cavernous hemangiomas showing dilated vascular channels with flattened endothelium.

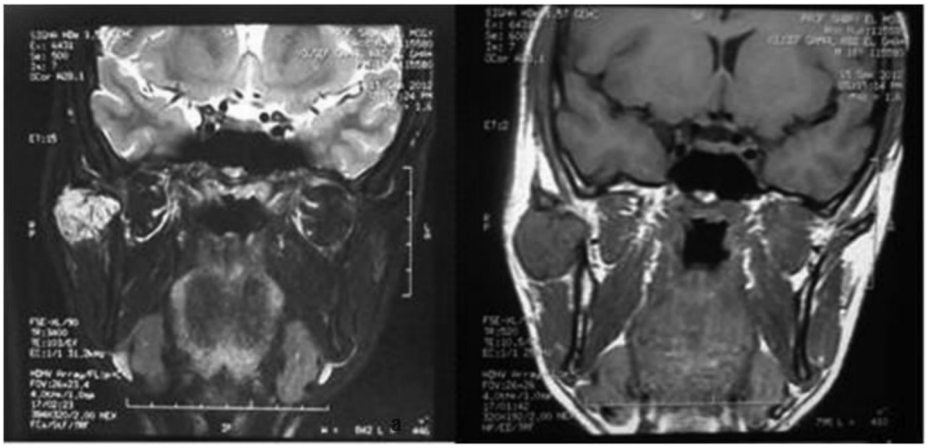

Fig. 4. MRI of a hemangioma in the superior part of the parotid gland. Note the strong signal produced by the tumor on T1 (a) weighted images, the weak signal produced on fat suppressed images (b), and the clearly-defined margin of the tumor. Note the extension of the mass to the condylar head.

are rare and no confirmative non-invasive investigation exists [9]. Plain X ray may show multiple calcified phelebolith while ultrasonography may reveal heterogeneous, hypoechoic lesions with calcified phleboliths. CT scan shows tumor with enhancing quality of blood vessels and will confirm the location of the tumor, as seen here in the present case, the tumor presented in the superficial lobe. However the soft tissue density of the tumor, its homogeneity, and any areas of extension are best demonstrated by MRI.

Magnetic resonance imaging demonstrates hyperintensity on T1 weighted images and isointensity with muscle on T2 weighted images. In addition, MRI scan of this hemangioma clearly showed the margin of the tumor and revealed the distinct outline of the lesion which enabled us to readily distinguish the tumor from the surrounding tissue. In the present case the images suggested a diagnosis of parotid hemangioma with medial extension to the condylar head, which proved true at surgery (Fig. 4). We thus believe that MRI is highly useful, perhaps even necessary, in diagnosing tumors of the head and neck.

Treatment modalities of haemangiomas include laser, cryotherapy, embolization and corticosteroids. The treatment of choice for this case was surgical resection followed by regular follow up. From oncosurgical, cosmetic and functional standpoints, the standard parotidectomy approach (face lift incision) or ‘S-incision’ was the selected approach (Fig. 5). However, Xie et al. [10] introduced a minimally invasive endoscopic approach via a small preauricular incision performed on 5 patients with benign APG tumors and no postoperative complications and recurrences were found.

Fig. 5. Intraoperative photograph showing the lesion and its extension to the condylar head.

Cupped out resorption of the condylar cortex indicates the long standing non invasive nature of this hemangioma. The superior border of the parotid gland (usually the base of the triangle) is closely molded around the external acoustic meatus and the temporomandibular joint, the lesion was closely attached to the lateral surface of the disc, so sharp dissection was done and no decision was taken to remove the condylar head. The specimen of the excised soft tissue cavernous hemangioma was firm because of fibrosis. The mass had a slightly blue color with a smooth surface. A cavernous hemangioma in the parotid is a low flow hemangioma and therefore there was no risk of a significant bleeding at surgery.

4. Conclusion

A unique case of cavernous hemangioma arising in the upper part of parotid gland is presented. Furthermore, our findings indicate that the efficacy of MRI was not limited to its use in making a diagnosis, but extended to the determination of the extension of the lesion to the temporomandibular joint, and its ability to aid selecting an appropriate surgical approach and the extent of dissection.

Competing interests

The authors declare that they are not in any conflict of interest.

Funding

Neither author nor any member of their families received any material or financial gain or personal advancement in the production of this manuscript.

Appendix A. Supplementary data

Supplementary data related to this article can be found at http://dx.doi.org/10.1016/j.tdj.2014.06.005.

References

[1] Carlson ER, Ord RA. Textbook and color atlas of salivary gland pathology. Diagnosis and management. USA: Wiley-Blackwell; 2008. pp. 19–67.
[2] Williams PL. Gray's anatomy. 38th ed. Oxford: Blackwell; 1995.
[3] Gosain AK, Yousif NJ, Madiedo G. Surgical anatomy of the SMAS: a reinvestigation. Plast Reconstr Surg 1993;92:1254–63.
[4] Marx DS, Robert E. Oral and maxillofacial pathology: a rationale for diagnosis and treatment. 1st ed., vol. 528. Hong Kong: Quintessence; 2003.
[5] Bater MC. An unusual case of preauricular swelling: a giant parotid sialoeele. Int J Oral Maxillofac Surg 1998;27:125–6.
[6] Gürbüzler L, Ceylan A, Yılmaz M, Vural C. Castleman's disease in the parotid gland: a case report. Kaohsiung J Med Sci 2010;26:444–7.
[7] Yadav JS, Chouhan M, Mody R, Bakhsi J. Heamangioma of submandibular gland–a rare case report entity. Int J Pediatr Otorhinolaryngol 2011;6:189–91.
[8] Nagao K, Matsuzaki O, Shigematsu H, Kaneko T, Katoh T, Kitamura T. Histopathologic studies of benign infantile hemangioendothelioma of the parotid gland. Cancer 1980;15:2250–6.
[9] Kimura Y, Ishikawa N, Goutsu K, Kitamura K, Kishimoto S. Lipoma in deep lobe of the parotid gland: a case report. Auris Nasus Larynx 2002;29:391–3.
[10] Xie L, Zhang D, Lu MM, Gao BM. Minimally invasive endoscopic-assisted resection of benign tumors in the accessory parotid gland: 5 case studies. Head Neck 2012;34(8):1194–7.